

Eosinophilic Pneumonia in a Patient with Bronchial Myiasis

Case report and literature review

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الالتهاب الرئوي اليوزيني لدى مريض يعاني من النعف القصبي في عمان تقرير حالة ومراجعة الأدبيات

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ABSTRACT: Pulmonary myiasis is an unusual form of myiasis in humans and has been recently identified as a cause of eosinophilic pneumonia. We report the case of a 13-year-old Omani boy who presented to the Royal Hospital, Muscat, Oman, in October 2014 with respiratory distress. Bronchial aspirates revealed features of eosinophilic pneumonia. Possible larvae identified in the cytology report, a high immunoglobulin E level and the patient history all indicated bronchial myiasis. The patient was treated with steroids and ventilation and has since been disease-free with no long-term side-effects. To the best of the authors' knowledge, this is the first case of bronchial myiasis in Oman.

Keywords: Myiasis; Eosinophilic Pneumonia; Case Report; Oman.

المخلص: داء النعف الرئوي هو شكل من الأشكال غير الاعتيادية من داء النعف في البشر وتم تعريفه مؤخرًا كأحد الأسباب للالتهاب الرئوي اليوزيني. فقد قمنا بتحرير حالة لطفل يبلغ من العمر 13 عامًا بعد قدومه للمستشفى السلطاني بمسقط في أكتوبر من عام 2014 وكان يعاني من ضيق في التنفس حيث أدخل المستشفى، وكشفت عينات الغسيل الرئوي أن الطفل يعاني من الالتهاب الرئوي اليوزيني. وبينت عينات الغسيل الرئوي، وارتفاع نسبة الغلوبين المناعي E والتاريخ المرضي للمصاب أن المريض كان يعاني من داء النعف القصبي. عولج المريض باستخدام التنفس الاصطناعي وعقار السيرويد وتعافى تماما من المرض بدون أي اعراض سلبية. وتعتبر هذه الحالة من داء النعف القصبي هي الأولى المسجلة في سلطنة عمان.

مفتاح الكلمات: داء النعف: الالتهاب الرئوي اليوزيني: تقرير حالة: عمان.

MYIASIS IS THE INFESTATION OF THE vertebrate tissues with fly larvae. Most human cases of myiasis in Oman are ophthalmomyiasis caused by *Oestrus ovis* (sheep bot fly) of the *Oestridae* family.^{1,2} Tracheopulmonary myiasis is very rare with only a few cases reported worldwide.³⁻⁹ This case report presents a 13-year-old Omani boy with an unusual case of bronchial myiasis resulting from the inhalation of mature larvae which caused a severe pulmonary reaction. A literature review of all reported cases of tracheopulmonary myiasis is also presented. Myiasis due to *O. ovis* remains a rare cause of pulmonary disease and is a newly described cause of eosinophilic pneumonia in humans.⁹

Case Report

In October 2014, a previously healthy 13-year-old boy from a mountainous area of interior Oman reported inhaling a fly and experiencing a subsequent sudden coughing fit. Upon inspection by a relative, all that

could be seen was some white thready material at the back of his throat. He presented to the local health centre with a progressive cough and breathing difficulty. The patient was hypoxic with an oxygen (O₂) saturation level of 90% when breathing room air. He was wheezy and in severe respiratory distress. He received salbutamol nebulisation without noticeable improvement. He was transferred to a regional hospital on the same day where he was adequately managed for wheezing. However, the patient remained hypoxic, requiring 3 L of O₂ to maintain an O₂ saturation level above 92%. As the patient was not improving, he was referred the next day to the Paediatric Emergency Department at the Royal Hospital, a tertiary centre in Muscat, Oman, for an urgent bronchoscopy. On examination, he was found to be in severe respiratory distress, with wheezes on the right side of his upper chest and minimal air entry. He was subsequently moved to the Paediatric Intensive Care Unit (PICU).

In the PICU, the patient was alert and conscious but still struggling to breathe; he needed 5 L of O₂

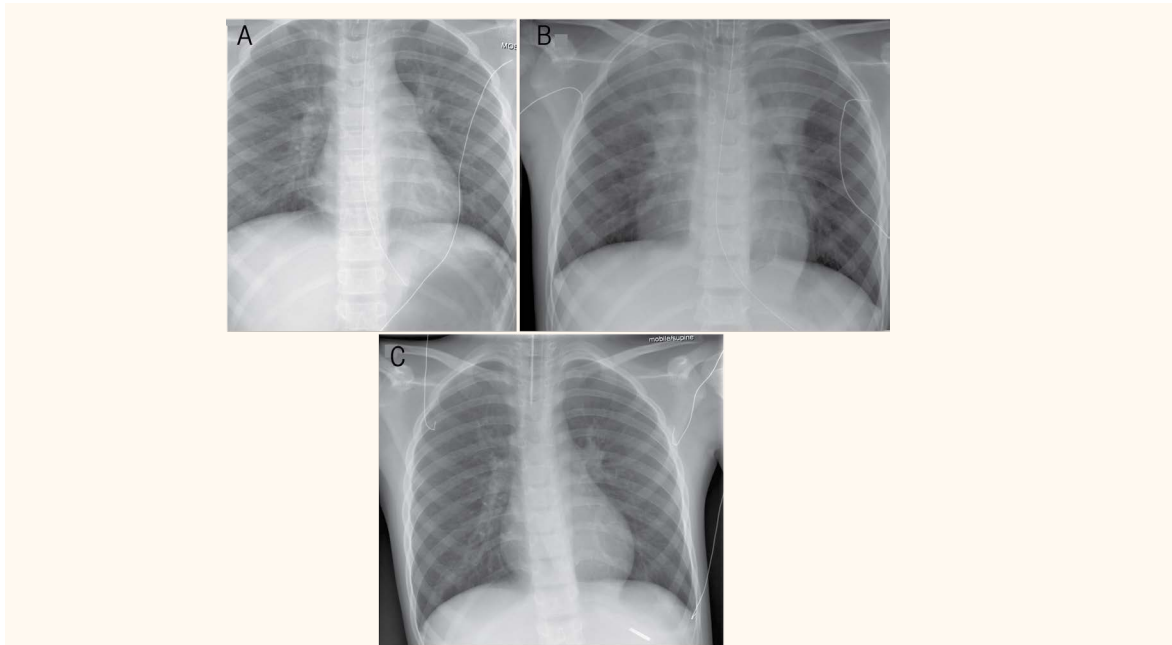


Figure 1A–C: Portable post-intubation supine chest X-rays of a patient with pulmonary myiasis and eosinophilic pneumonia at (A) admission, (B) two days later and (C) four days after high-frequency oscillatory ventilation (HFOV). Note the worsening pneumonia with bilateral involvement and the improvement after HFOV.

administered via a face mask and could only maintain an O_2 saturation level of 90%. Due to persistent hypoxia, the patient was intubated with a 6.5 size cuffed tube and mechanically ventilated for 10 days with synchronised intermittent mandatory ventilation with volume control. A direct laryngoscopy did not reveal any obvious vocal cord oedema. Flexible bronchoscopy showed oedema and thick secretions in the right upper lobe and superior segment of the right lower lobe. However, no obvious foreign body was isolated. The patient developed anaphylactic shock and was treated with a 12-hour adrenaline infusion with good response. A repeat flexible bronchoscopy after two days revealed thick whitish mucous casts but no foreign body. This time, involvement of the left side was also noted. X-rays showed an initial worsening of the pneumonia followed by gradual improvement after four days of high-frequency oscillatory ventilation (HFOV) [Figure 1]. Post-extubation, he did not require any further O_2 .

Cytology of the bronchial aspirate revealed eosinophils in scattered clusters. Many neutrophils, macrophages and lymphocytes were also admixed with a thick mucus. Occasional small oval structures (possibly larvae) were identified [Figure 2] along with a few Charcot-Leyden crystals. There were no bacteria or fungi isolated in the culture and a complete blood count did not show any eosinophilia. The presence of the eosinophilic clusters and Charcot-Leyden crystals in the cytology report suggested a diagnosis of eosinophilic pneumonia. The presence of possible larvae in the cytology report, a

high immunoglobulin E level (897 IU/mL) and the patient history all pointed towards a parasitic cause. The eosinophilic pneumonia was treated with 14-day courses of intravenous hydrocortisone and ceftriaxone, the former of which was then tapered off over the following seven days.

Discussion

Myiasis is caused by the invasion of human or animal tissues by fly larvae (maggots) and can be classified as accidental, facultative or obligate. Human cases are mostly accidental. In terms of the present case,

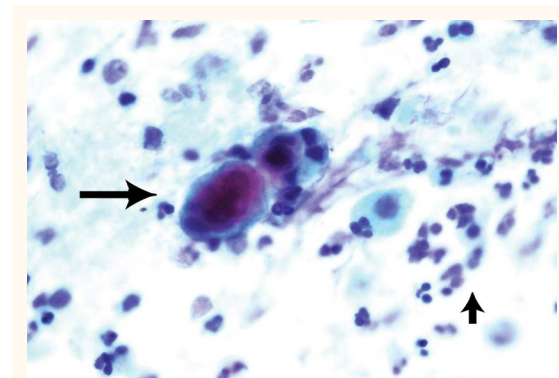


Figure 2: Papanicolaou stain at x400 magnification showing an amphiphilic structure (large arrow) consistent with an *Oestrus ovis* larva in a patient with pulmonary myiasis and eosinophilic pneumonia. Note the inflammatory infiltrate with a predominance of eosinophils (small arrow).

Table 1: Demographic and entomological characteristics of tracheopulmonary myiasis cases in the literature

Author and year of report	Patient age in years and gender	Location	Suspected causative organism	Involvement	Confirmatory findings	Larvae stage
Ahmed <i>et al.</i> ³ 1969	N/A	USA	<i>Gasterophilus</i> genus	Pulmonary	N/A	N/A
Scholten <i>et al.</i> ⁵ 1977	22 M	Canada	<i>Cuterebra fontinella</i>	Tracheal	Patient coughed up larvae	Instar 2
Komori <i>et al.</i> ⁷ 1978	19 M	Japan	<i>Megaselia spiracularis</i>	Pulmonary	Larvae on lung biopsy	Instar 3
Baird <i>et al.</i> ⁴ 1979	22 M	USA	<i>Cuterebra</i> subgenus	Bronchial	Patient coughed up larvae	Instar 2
Fraiha <i>et al.</i> ⁶ 1984	N/A	Brazil	<i>Alouattamyia baeri</i>	N/A	N/A	N/A
Choi <i>et al.</i> ⁸ 2002	84 F	Korea	<i>Lucilia</i> genus	Pulmonary	Larvae crawled out of patient's nose	N/A
Cornet <i>et al.</i> ⁹ 2002	60 F	USA	<i>Cuterebra</i> subgenus	Tracheal	Patient coughed up larvae	Instar 3
Present case	13 M	Oman	<i>Oestrus ovis</i>	Bronchial	Bronchial aspirates	N/A

N/A = not available; M = male; F = female.

the responsible organism for the myiasis was likely to be *O. ovis*, locally known as *dis'sa*, because the fly is known to lay its larvae during flight and the species is very common in the patient's home region. The eyes are usually affected in most cases of *O. ovis* involvement.^{1,2,10} Many people living in the same area as the patient, including the boy's father, had had a history of conjunctivitis and coughing.

O. ovis is found worldwide and is known to cause cavitary myiasis. Its hosts are sheep and goats and there is as yet no confirmation that *O. ovis* is able to complete its life cycle in humans. The female fly is active during the summer and early autumn, when it deposits as many as 500 already hatched first instar larvae. Direct contact between the fly and its host is not necessary for infestation.¹⁰ *O. ovis* is the most common cause of external ophthalmomyiasis and may cause many allergic manifestations, especially when found in the throat.^{11,12}

Tracheopulmonary myiasis is a very rare form of the disease; to date, only seven cases have been reported worldwide [Table 1].³⁻⁹ The geographical distribution of cases is varied (including the USA, Brazil, Japan and Korea) and no specific age group or gender is targeted. Causative organisms range from *Cuterebra fontinella*, *Megaselia spicularis*, *Alouattamyia baeri* and the *Lucilia* and *Gasterophilus* genera and *Cuterebrinae* subfamily. Although the likelihood of human infestation is rare, the severe medical complications and the varied worldwide distribution indicates the medical importance of this condition. As with the current patient, those suspected of having tracheopulmonary myiasis (i.e. those with

severe respiratory distress and hypoxia and living in fly-prevalent regions) should be urgently treated at a tertiary care centre.

Eosinophilic pneumonia is a condition describing an accumulation of eosinophils in the lungs. Known causes include certain medications, parasitic infections, cancer, environmental triggers and immunogenic factors. The prognosis is very good and patients usually respond well to steroid treatment. Parasites can cause eosinophilic pneumonia in three different ways: either by invading the lung, living in the lung as part of their life cycle or spreading to the lung through the bloodstream. Tapeworms, helminths and Japanese lung flukes are the most common parasites. Fly larvae infestation has recently been reported as a cause of eosinophilic pneumonia and is a very rare entity.⁹

A few cases of ophthalmomyiasis have been reported in Oman; however, only some of these were caused by *O. ovis*.^{1,2} There have been no reported cases of bronchial myiasis in Oman to date. The current case represents the first report of bronchial myiasis in Oman and the Middle East with eosinophilic pneumonia requiring mechanical ventilatory support, including HFOV, despite treatment with steroids.

Conclusion

Bronchial myiasis can be a life-threatening infestation and should be considered in the differential diagnosis of unexplained respiratory distress and hypoxia in individuals from fly-prevalent areas. Patients should be urgently referred to a tertiary centre for effective management and care. To the best of the authors'

knowledge, this is the first case of bronchial myiasis in Oman. The patient presented with eosinophilic pneumonia and the causative organism was likely *O. ovis*.

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